



Tumor-Like Cystic Endosalpingiosis in the Myometrium: A Case Report and a Review of the Literature

Myometriumda Tümör Benzeri Kistik Endosalpingiozis: Olgu Sunumu ve Literatürün Gözden Geçirilmesi

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ABSTRACT

Endosalpingiosis is a disorder of the Mullerian system characterized by benign glands lined with tubal type epithelium and involves the peritoneum, subperitoneal tissues and retroperitoneal lymph nodes. Myometrial endosalpingiosis is very rare. A 44-year-old female presented to the hospital complaining of menorrhagia. Gynecological and radiologic examination revealed a pelvic mass 7.5 cm in diameter, suggestive of a leiomyoma with secondary cystic changes. The gross examination showed an intramural large cyst within the right fundic wall of the uterus. Microscopically, the cyst consisted of multiple micro-macro cysts lined with benign tubal epithelium. With these findings, the diagnosis was cystic endosalpingiosis. In this report, the clinicopathologic features of a cystic endosalpingiosis case located in the myometrium was presented with a review of the literature.

Key Words: Cystic endosalpingiosis of the uterus, Uterus, Cysts

ÖZ

Endosalpingiozis, periton, subperitoneal dokular ve retroperitoneal lenf nodlarını tutan tubal tip epitel ile döşeli benign glandlarla karakterize bir Müllerian sistem bozukluğudur. Myometrial endosalpingiozis çok nadirdir. Menoraji şikayeti ile hastaneye başvuran 44 yaşındaki kadın hastanın jinekolojik ve radyolojik muayenesinde sekonder kistik değişiklikler içeren, kuvvetle leiomyomu düşündürülen 7,5 cm çapında pelvik kitle saptandı. Makroskopik incelemede, uterin fundus sağ duvarında intramural büyük bir kist izlendi. Mikroskopik olarak; kist, benign tubal epitel ile döşeli, çok sayıda makro ve mikro kistlerden oluşmakta idi. Bu bulgular ile kistik endosalpingiozis tanısı kondu. Bu makalede myometriuma lokalize kistik endosalpingiozis olgusunun klinikopatolojik özellikleri literatür gözden geçirilerek sunulmaktadır.

Anahtar Sözcükler: Uterusun kistik endosalpingiozisi, Uterus, Kist

INTRODUCTION

Endosalpingiosis is a non-neoplastic process characterized by the presence of tubal type epithelium-lined glandular structures (1). It is derived from the Mullerian system and is seen in the pelvic and lower abdominal peritoneum. Colon, bladder, appendix, paraaortic and pelvic lymph nodes are the sites specified in the case reports (2-4). Patients are almost always asymptomatic and it very rarely presents as a large cystic mass known as florid cystic endosalpingiosis (5). Endosalpingiosis is generally associated with serous neoplasms of the ovary (1). Here we present a case with a purely intramural uterine cystic endosalpingiosis, a benign serous tumor in the right ovary and bilateral follicular salpingitis. This is the first case with intramyometrial involvement in the English literature to our knowledge.

CASE REPORT

A 44-year-old woman presented with a history of menorrhagia for six months. Physical examination revealed

a pelvic mass, confirmed on the ultrasound as arising from the uterus. Total abdominal hysterectomy and bilateral salpingo-oophrectomy (TAH+BSO) was performed. On gross examination, each fallopian tube was measured 4x1 cm and there was a 6 cm cyst on the right ovary and 3 cm cyst on the left ovary. Intraoperative evaluation of both ovaries was reported as benign. The uterus was 10x9x4 cm in size and revealed an intramural biloculated cyst in the fundus. The cyst contained serous fluid and measured 8 cm (Figure 1). Microscopic evaluation revealed florid cystic spaces of variable size and gland-like abortive structures lined with ciliated cuboidal serous epithelium (Figures 2,3). The stroma between the cystic/glandular spaces was muscular, identical to myometrial smooth muscle. Neither endometrial stromal differentiation nor recent/old hemorrhagia or endometrial glands was noted. Both fallopian tubes had shortened and fibrotic plicae. Plical fusion in the tubes was caused by follicle-like structures and mixed stromal inflammatory infiltration that was diagnosed as a follicular salpingitis.

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The right ovarian cyst was a benign serous tumor covered by a single layer of flattened epithelium. The left ovary also revealed a follicular cyst.

DISCUSSION

Endosalpingiosis refers to the presence of tubal epithelium outside the uterine tube and it is generally considered an incidental finding (5). It is not considered in the differential diagnosis at the first glance, particularly from the clinical aspect, both because of its rarity and because it is overshadowed by endometriosis that is seen frequently in



Figure 1: The uterine cut surface with cyst formation without relation to the endometrium.

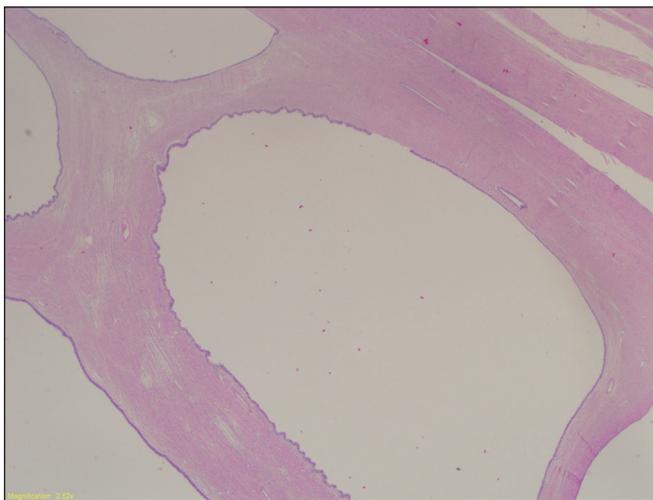


Figure 2: Multiple cysts of variable sizes lined with benign epithelium are seen (H&E x40).

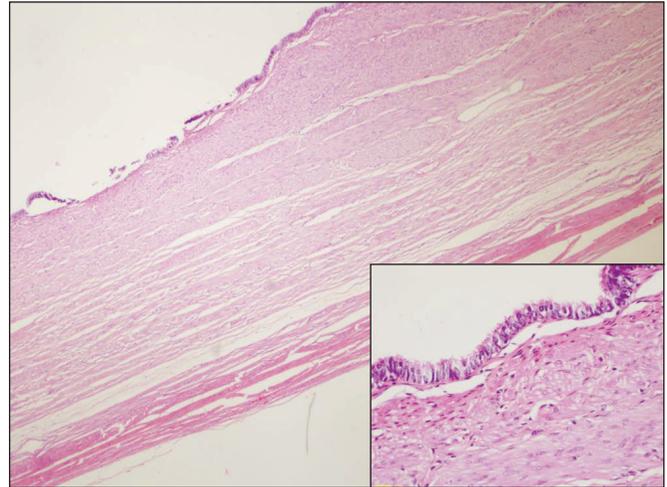


Figure 3: Cystic structure located within the myometrial fibers. A cut section of the thinnest part of the myometrial wall is shown (H&E x10). Inset: The cysts are lined by ciliated tubal epithelium (H&E x40).

gynecology practice. The symptomatology is not specific so it is usually misinterpreted (6). The current case's complaint was menorrhagia, similar to the cases reported before.

Many extragenital locations have been reported in the literature such as peritoneum, subperitoneal tissue, colon, appendix, umbilicus, and lymph nodes (2,4). The examples of tumor-like cystic endosalpingiosis were localized to the female pelvis and most of those cases were reported to be derived from the serosal surface of the uterus and the ovary (1,6-11). The tumor-like cystic endosalpingiosis was located in a subserosal location with intramural involvement in only six cases previously published in the literature (1,5,12-14). The case presented here was different from the cases reported before with its feature of purely uterine-intramural cystic tumor-like endosalpingiosis without a serosal component as published by Çil et al. (15) (Table I).

Tumor-like cystic endosalpingiosis usually mimics other lesions such as leiomyoma, serosal cyst etc. (1, 9). The lesion mimicked leiomyoma in the current case as well. The differential diagnosis of endosalpingiosis includes cystic tumor-like lesions involving the uterus such as cystic adenomyosis with tubal metaplasia, adenomatoid tumor, and peritoneal inclusion cyst (5,6,8,12,13). Absence of endometrial stroma differentiates cystic adenomyosis from endosalpingiosis. Cystic adenomatoid tumors may present as a uterine cystic mass. They have multiple small ovoid or slit-like spaces lined with flattened and cuboidal non-tubal type epithelial cells admixed with smooth muscle fibers (5,8). They are usually located on the subserosal area of the female genital tract, sometimes with a myometrial invasion.

Table I: Summary of tumor-like cystic endosalpingiosis of the uterine cases reported in the literature

Case No	Author	Ref. No	Age	Symptoms	Localization	Gross Examination	Treatment
1	Clement	1	20	Incidentally in delivery	subserosal	Multiple cysts	Multiple biopsies
2	Clement	1	41	Menorrhagia, dyspareunia	subserosal and intramural	Cysts with smooth lining and clear fluid	TAH
3	Clement	1	43	Menorrhagia, pelvic pain	subserosal and intramural	Cysts with smooth lining and brownish fluid	TAH+BSO+pelvic lymphadenectomy for cervical cancer
4	Shim	5	54	Vaginal bleeding	Subserozal and intramural	Subserosal cystic mass	TAH+BSO
5	Rosenberg	6	50	Pelvic pain	Subserozal	Large cystic mass with yellow viscous fluid	Excision of the mass
6	Heatley	7	73	Abdominal swelling	Subserosal	Multiple solid nodules	TAH+BSO
7	Chang	8	45	Pelvic pain	subserosal	Pedunculated multilobular cystic mass	TAH
8	Taneja	9	40	Pelvic pain	subserosal	Large, soft, tender mass	TAH+BSO
9	Bryce	10	25	Incidental	subserosal	Multicystic lesion	Multiple biopsies
10	Lee	11	54	Incidental	subserosal	Two oval cystic and solid masses	TAH+BSO
11	Fukunaga	12	51	Pelvic pain	subserosal and intramural	Multilobular masses	TAH+BSO
12	Kajo	13	50	Palpable mass in the pelvis	subserosal and intramural	Mass with multiple cysts	TAH+BSO
13	Youssef	14	49	Menorrhagia	subserosal and intramural	Pedunculated cystic mass and multiple cysts in uterine serosa and both ovaries	TAH+BSO
14	Çil	15	45	Menorrhagia and pelvic pain	intramural	Intramural, serous fluid filled cystic mass	TAH+BSO
15	Current case		44	Menorrhagia	intramural	Large cystic mass	TAH+BSO

TAH: Total abdominal hysterectomy, **BSO:** Bilateral salpingo-oophorectomy.

However, there was no evidence of serosal involvement in our case and immunohistochemistry would be helpful in the differential diagnosis otherwise.

The etiology and the histogenesis of cystic endosalpingiosis is not clear, but may be related to inflammatory conditions, previous adnexal surgery or peritoneal mesothelial metaplasia depending on the growth factor stimuli or steroid hormones (13). There is some evidence that it might be a secondary müllerian system pathology with mullerianosis; such as lymphadenectomy specimens resected due to ovarian serous carcinoma usually

containing endosalpingiosis, endosalpingiosis cases almost always being accompanied by a serous cystic neoplasm, and urinary bladder endosalpingiosis (1,16). Our case had follicular salpingitis that might support the idea of a possible relationship between an inflammatory process and this entity.

In conclusion, endosalpingiosis rarely presents with such large cystic changes and striking macroscopic findings. Awareness of the existence of this rare lesion will prevent an incorrect diagnosis.

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